

Odontogenic Cyst of 21st Century: Glandular Odontogenic Cyst-Case Report

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Abstract:

Glandular Odontogenic Cyst" (GOC) is a rare developmental cyst which is histopathologically unusual type of odontogenic cyst. It is a well-known entity, because of the facts that it exhibits a propensity to recur and has aggressive potential. Recurrence rate of Glandular Odontogenic Cyst ranges from 21% to 55%. Microscopically it resembles central mucoepidermoid carcinoma. Moreover, some of its microscopic features are found in dentigerous, botryoid, radicular & surgical ciliated cyst. This paper has well-elaborated algorithm for histopathological evaluation. In spite of being rare, it has been noted to have aggressive potential, high incidence of cortical perforation & a relatively high rate of recurrence. Hence, correct diagnosis is challenging & clinically very important. This case report highlights two cases of Glandular Odontogenic Cyst one occurring in the maxillary posterior which is rare and unusual and other in the maxillary anterior region which is the second most common site of Glandular Odontogenic Cyst. Clinical & radiological presentation of both the cases did not lead to correct diagnosis. Hence, histopathological evaluation was the ultimate tool for the diagnosis.

Keywords: Glandular Odontogenic Cyst, Dentigerous Cyst, Cyst Enucleation.

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Introduction:

Glandular Odontogenic cyst (GOC) is a rare developmental cyst.¹ Padayache and Van Wyk in 1987, reported the first two cases of this cyst as "Sialo-Odontogenic Cyst".² Gardener et al. in 1988, established it as a distinct entity and coined the term "Glandular Odontogenic Cyst." GOC was included in the list of histologic typing of an odontogenic tumour in the year 1992 by the World Health Organization (WHO) with the term GOC or Sialo-Odontogenic Cyst.

The most common site of occurrence of GOC is the anterior mandible.³ GOC has a slight male predilection.³ on radiographic examination it shows well-defined, unilocular or multilocular radiolucency having scalloped margins. Histopathologically, GOC shows characteristics features that include non-

keratinized stratified squamous lining epithelium, focal thickening (plaques) within the lining, eosinophilic cuboidal or columnar cell (Hobnail cells) that may be ciliated, papillary projections of epithelium, mucous cells, interepithelial gland-like structures and inflammatory cells in the sub epithelial connective tissue.⁴

It has two clinical features: it has a "high recurrence rate," and it displays an "aggressive growth potential".⁵ Definitive diagnosis of GOC has a complex and frequently non-specific histopathology. GOC has a histopathological resemblance to a well-differentiated (low grade) mucoepidermoid carcinoma, requiring entirely different treatment and surgical intervention.⁶ GOC is an recently recognized clinical entity, so it needs to be diagnosed properly to check for its potential aggressive behaviour and a tendency to recur. Literature has recommended more

stringent criteria for the histopathological diagnosis of GOC. If any degree of specificity occurs in clinical and radiological features, then it should be identified and be recruited to refine further the diagnosis.

Curettage and Enucleation are considered to be the first line of treatment, although some author prefers marginal resection due to its tendency to recur after curettage or Enucleation.

CASE REPORT:

CASE 1:

A female patient of 45years old, reported to the outpatient Department of Oral & Maxillofacial Surgery at M.I.D.S.R. Dental College, Latur with the chief complaint of swelling in the upper right posterior region of the jaw since last one year. The patient was asymptomatic except for mild pain for the last one month. Initially swelling was of pea-sized, which gradually increased to approximately about 3 x 2 x 1 cm in dimensions. An extra oral examination revealed no significant clinical finding. Intraoral examination revealed an oval- shaped swelling over the right palatal region extending antero-posteriorly from 16 to 18 and medially from the mid palatal raphe till the gingival margin of the associated teeth i.e. 16, 17 & 18 laterally. Swelling had well-defined borders, and overlying mucosa appeared reddish pink with no evidence of pus discharge. On palpation, the swelling was not fluctuant, non-tender & firm in consistency. Associated teeth, i.e., 16, 17 & 18 were vital. Clinically it appeared as mucoepidermoid cyst, but on fine needle aspiration, it yielded serous brownish red colored fluid suggestive of the cystic lesion and hence, mucoepidermoid carcinoma was ruled out.

A panoramic radiograph revealed well-defined, unilocular radiolucency with sclerotic borders involving right maxillary sinus. Interspersed within the radiolucency were the radiopacity suggestive of haziness in the right maxillary sinus. The radiolucency extended from 16 to 18. Root resorption associated with 16, 17 & 18 was appreciable (**Fig.1**).



Figure 1: Panoramic image is showing well-defined, unilocular, radiolucency with sclerotic borders.

The lesion was treated conservatively with Enucleation and curettage under general anaesthesia. The specimen was then sent for histopathological examination.

Histopathological examination revealed cystic cavity lined by glandular epithelium with epithelial crypts present. Mucous cells were seen interspersed within the lining of the epithelium. The cystic lining was fibrous with inflammatory cells (**Fig. 2**).

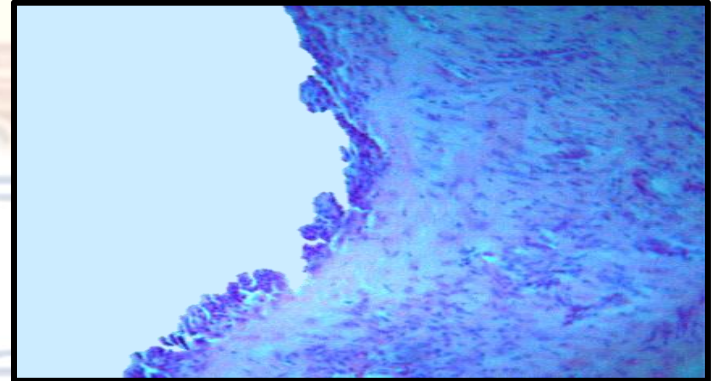


Figure 2: Photomicrograph is revealing cystic lining exhibiting variable thickness of epithelium with epithelial crypts and mucous cells.

Based on these finding histopathological diagnosis of GOC was made.

CASE 2:

A 30 years old male patient reported to the outpatient Department of Oral & Maxillofacial Surgery at M.I.D.S.R. Dental College, Latur with the chief complaint of swelling and pain in the upper anterior region of the jaw since last six months. No

significant findings were noted extra orally. Intraorally, over the maxillary anterior region of the palate, oval-shaped swelling with defined borders was present. The swelling was approximately about 3 x 2 cm in dimension over mid-palatal raphe. The lesion was approximately 2 cm away from the gingival margin of the maxillary anterior teeth. Overlying mucosa appeared normal without any evidence of pus discharge. It was non-fluctuant, tender and firm in consistency on palpation. On performing vitality test, 11 & 12 were non-vital. 11 was root canal treated. A considerable amount of teeth displacement with 13, 12, 11 & 21 was present. Fine needle aspiration yielded yellowish brown colored fluid.

Panoramic radiograph showed a well-defined, unilocular radiolucency with corticated borders, extending from mesial of 15 to mesial of 22. Teeth displacement with 13, 12, 11 & 21 was appreciable. No root resorption was present with any involved teeth (**Fig.3**).



Figure 3: Panoramic image is showing well-defined, unilocular radiolucency with corticated borders

The Lesion was treated conservatively with Enucleation and curettage under general anaesthesia. The specimen was then sent for histopathological examination. Histopathologically it revealed cystic lesion with luminal epithelium and surrounding connective tissue. Throughout the lining of the epithelium, glandular-like structures and mucous cells were present. GOC was the histopathological diagnosis made.

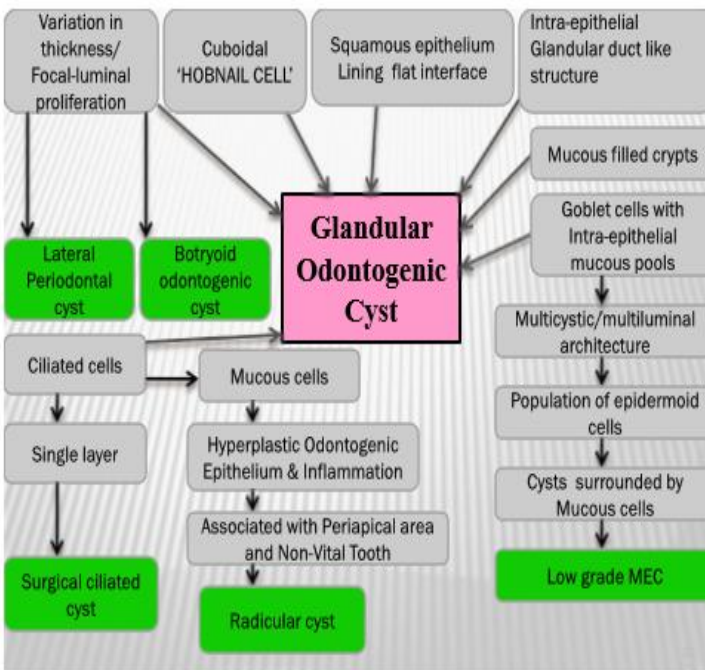
Discussion:

GOC accounts for 0.012% to 1.3% of all the jaw cysts, and its prevalence is 0.17%.⁶ The most common site of occurrence is the mandible (85%), especially in the anterior region, followed by the anterior of the maxilla. The posterior region of the maxilla is a rare area of occurrence for GOC. In case 1, the lesion was present in the maxillary posterior region which is a very rare finding.

GOC does not have specific or pathognomonic radiographical features⁷. It may present as a multilocular or unilocular radiolucency with well-defined borders. The diagnosis based on clinical and radiological examination is practically impossible. Only the histopathological examination allows for diagnosis of the cyst.⁷

Under general anaesthesia, both the cases were treated with Enucleation and curettage. The diagnosis of GOC was done after Enucleation and histopathological study of the lesion. Since it has high recurrence rate, aggressive surgical management would have been our line of treatment. Hence, follow up with radiological evaluation of the patient was done every six months. The treatment of GOC ranges from a conservative approach (Enucleation, marsupialisation, curettage with or without peripheral osteotomy, curettage with adjuvant Carnoy's solution, or cryotherapy) to marginal resection and segmental resection.⁸ Some authors prefer marginal and segmental resection due to a tendency of the cyst to recur after conservative treatment. In our cases, both the clinical and radiographical finding did not co-related with the actual nature of the cyst demonstrated in the literature, thereby leading to diagnostic and treatment dilemma.

Following is the well-elaborated algorithm showing a histopathological differential diagnosis of GOC which lead us to the correct diagnosis



Conclusion:

To conclude, the careful histopathological examination is needed to diagnose GOC as the clinical and radiological findings are overlapping with lateral periodontal, BOC, residual, and radicular cyst. A careful, long-term follow up is advocated, because of its aggressive behaviour and the tendency for recurrence.

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